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Development of a Multi-dimensional Scale for PDD and ADHD

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Abstract

A novel assessment scale, the Multi-dimensional Scale for Pervasive developmental disorder (PDD) and Attention-deficit/hyperactivity disorder (ADHD) (MSPA), is reported. Existing assessment scales are intended to establish each diagnosis. However, the diagnosis by itself does not always capture individual characteristics or indicate the level of support required, since inter-individual differences are substantial and co-morbidity is common. The MSPA consists of 14 domains and each domain is rated by a nine-point quantitative scale. The clinical and behavioral features are projected onto a radar-chart, which facilitates understanding of the disorders both by the patients themselves and by those in their surroundings. We assessed 179 patients and analyzed features by six diagnostic subgroups, which showed relationships between features and diagnoses. The inter-rater reliability was satisfactory.

Keywords: Pervasive developmental disorder, ADHD, autism, assessment, chart

1. Introduction

Pervasive developmental disorder (PDD) and attention-deficit/hyperactivity disorder (ADHD) belong to the class of neurodevelopmental disorders. The former is characterized by severe and pervasive impairment in several areas of development, which may include reciprocal social interaction skills, communication skills, and the presence of stereotyped behavior, interests, and activities, according to the Diagnostic and Statistical Manual of Mental Disorders - Fourth Edition - Text Revision (DSM-IV-TR) (American Psychiatric Association, 2000). The latter is characterized by hyperactivity, inattention and impulsivity. However, PDD patients often have symptoms of ADHD (Jensen, Larrieu, & Mack, 1997; Yoshida & Uchiyama, 2004; Frazier & Youngstrom, 2006; Lee & Ousley, 2006; Sinzig, Walter, & Doepfner, 2009) and vice versa (Nijmeijer et al., 2009; Kochhar et al., 2010), although diagnostic criteria do not overlap (Ghanizadeh, 2010). Also, genetic linkages between these disorders have been reported (Yamagata et al., 2002; Smalley et al., 2002; Ogdie et al., 2003; Bakker et al., 2003; Lichtenstein, Carlström, Råstam, Gillberg, & Anckarsäter, 2010). In clinical practice, the differential diagnosis between ADHD and a milder subtype of PDD, PDD Not Otherwise Specified (PDDNOS), is sometimes difficult, because the criteria have not been so formulated as to be useful in this aspect of differential diagnosis, and

because it is often the case that a patient has symptoms of PDD as well as ADHD (Nijmeijer et al., 2008).

In addition, these patients often demonstrate clumsiness (Pitcher, Piek, & Hay, 2003; Strum, Fernell, & Gillberg, 2004; Dewey, Cantell, & Crawford, 2007; Pan, Tsai, & Chu, 2009; Staples, & Reid, 2010). Gillberg and Gillberg (1998) proposed the concept of DAMP (deficits in attention, motor control, and perception), which is an overlapping condition of ADHD and developmental coordination disorder (DCD). Moreover, PDD and ADHD patients often suffer from several other symptoms, such as sleep problems (Richdale & Schreck, 2009), sensory abnormality (Harrison & Hare, 2004; Leekam, Nieto, Libby, Wing, & Gould, 2007; Baron-Cohen, Ashwin, Ashwin, Tavassoli, & Chakrabarti, 2009; Lane, Young, Baker, & Angley, 2010) and learning or executive dysfunction (Nijmeijer et al., 2008).

Thus, many of the characteristics of PDD, ADHD and DCD are not specific to one diagnosis. The combination of these clinical characteristics has a wide inter-individual variation. Furthermore, the degree of dysfunction in each domain also varies from person to person. Therefore, the diagnosis alone may neither represent the entire profile of characteristics nor indicate the support that an individual patient may need in life. In addition, the diagnosis itself is very time-consuming because of the

amount of information that must be gathered. Consequently, only a fraction of the patients who need special care obtain specialized assessment (Russell, Ford, Steer, & Golding, 2010). Furthermore, not only before but also after diagnosis, these patients and their families face numerous struggles to receive appropriate support for all areas of disability and deficit (Whitman, 2004). Therefore, we developed a Multi-dimensional Scale for PDD and ADHD (MSPA) to describe their symptom profiles comprehensively and guide them to the specific support needed more directly. It consists of 14 domains of clinical and behavioral features including five core features of PDD, three of ADHD, two of DCD, and the four problem areas of sensory, sleep, learning, and language development.

2. Materials and methods

Ethical approval for the study was obtained from the Ethics Committee at Kyoto University Hospital. We conducted the study according to the Ethical Guideline for Epidemiological Research by the Japanese Ministry of Health, Labour and Welfare.

2.1. *Participants*

179 patients with PDD or ADHD were evaluated by psychiatrists who are experienced in examining these disorders. They visited psychiatrists for a diagnosis and

a professional assessment between September 2006 and July 2010. The diagnosis was based on the criteria of DSM-IV-TR. We measured IQ (intellectual quotient) by Wechsler Adult Intelligence Scale Third Edition (Wechsler, 1997) for patients above 17 years old or by Wechsler Intelligence Scale for Children Third Edition (Wechsler, 1991) for patients from 6 to 17 years old, or DQ (developmental quotient) by Kyoto Scale of Psychological Development for patients under 6 years old (Ikuzawa, Matsushita, & Nakase, 2001). We divided them into six diagnostic groups: autistic disorder with mental retardation (Autism with MR) (IQ or DQ < 70), autistic disorder without mental retardation (Autism without MR), Asperger's disorder, PDDNOS, combined-type ADHD, and inattentive-type ADHD. We excluded cases above 50 years old because of the difficulty in obtaining information from infancy, and also excluded cases with complications such as deafness. Consequently, the age of the subjects was 14 ± 10 (mean \pm standard deviation) years old, and the range was 3 - 49 years old. The profiles of the participants are presented in Table 1. Differences among groups were not seen in gender or age by one-way ANOVA. Autism with MR had significantly lower scores than each of the other groups in FIQ, VIQ and PIQ, as expected from the diagnostic definitions. No significant differences were seen between other pairs.

[place Table 1 about here]

2.2. Measures

We extracted 14 domains of clinical and behavioral features in PDD and ADHD patients: five from PDD features (communication, sociality, emotion, restricted interests/behaviors, stereotyped/repetitive motion), two from DCD (gross motor, fine motor), three from ADHD (hyperactivity, inattention, impulsivity), and four other symptom areas where these patients often suffer (sensory, sleep cycle, learning, and language development). We also formulated the criteria of a nine-rank scale in each domain according to the degree of difficulties in life. The anchor-points throughout the domains are as follows: 1: no sign; 2: somewhat but no need to support; 3: special needs by supervisors in groups; 4: special needs by everyone in groups; 5: still difficult even with full-support in groups and special needs in individual life; and 1.5, 2.5, 3.5 and 4.5 if the conditions are between the adjacent scores. The further criteria for each domain were formulated through repeated meetings among child psychiatrists (22 times in total) and opinions by other professionals. Symptom severity was defined using the concepts of DSM-IV-TR in the domains other than sleep cycle. With respect to learning, we used the concept of learning disorder. We gathered information on behaviors from birth until the present from patients, parents and teachers as much as possible by records and by interview. We evaluated the characteristics of the person assuming an average social

environment. If the person was in a very specific social environment, we attempted to exclude the influence of it in our ratings. Similarly, we attempted to exclude the influences of physical diseases or handicaps from our ratings. If the condition was unstable or information was inadequate, we rated the subjects after gathering sufficient information. Most importantly, as we assessed subjects with a range of ages using age-nonspecific measures, the rating was done in reference to normal development. Thus, the knowledge of normal development is an essential requirement for the raters.

Six patients could not remember their language development. Also, we could not score the learning ability in 19 children under school age, especially in the MR group.

2.3. Radar-chart representation

We used a radar-chart representation to visualize the entire profile of 14 domains (Figure 1). The place of each domain was arranged so that adjacent domains were related. PDD symptoms are at the upper right, domains related to motor are at the lower right, ADHD symptoms are at the lower left and others are at the upper left. With this technique, we could visually grasp the characteristics of subjects at a glance.

2.4. Statistics

To evaluate inter-rater reliability, four trained psychiatrists rated 20 cases

independently. The intraclass correlation coefficient (ICC) for each domain was calculated using SPSS 17.0.

Scores of 179 patients on each domain were also analyzed by SPSS 17.0. We compared the scores among the six groups described above using one-way ANOVA, and then we conducted post-hoc tests by the Tukey method.

3. Results

The scoring took about 15 minutes when adequate information was available.

[place Figure 1 about here]

3.1. Scoring examples

Three examples of the scored charts are shown in Figure 1a. Case 1 (blue in Fig. 1a) was diagnosed as Asperger's disorder because he did not act as a member of his social group and his interests were very restricted. However, the chief complaint was his going out from the classroom. He also had inattention, that is, ADHD-like symptoms. Patients of this pattern tend to be regarded as ADHD at a glance. The diagnosis of Case 2 (pink in Fig. 1a) was Asperger's disorder, same as Case 1, but the behavioral features were quite different. She was too clumsy to attend physical education class and the chief complaint was repetitive motion. Case 3 (green in Fig. 1a) was diagnosed as PDDNOS

and was hypersensitive to noise and touch. He could not stay in the classroom because of noise. Also, he had disabilities in reading and writing.

3.2. Reliability

The inter-rater reliabilities are shown in Table 2 in each domain. The mean and standard deviation was 0.933 ± 0.055 across domains, ranging 0.834 to 0.983, and was adequately high among the trained psychiatrists participating in the study.

[place Table 2 about here]

3.3. Feature Analysis

We showed mean scores per group in each domain on a radar-chart (Figure 1b). In general, inattention was present in all the groups. Group variances by one-way ANOVA were significant in the domains other than sleep cycle. We carried out post-hoc tests and show the results in Figure 2. We also calculated the percentages of patients at clinical level (score of 3 or above) in each group by domains (Table 3).

[place Figure 2 and Table 3 about here]

As a reflection of the diagnostic criteria, all the patients in the two autism and Asperger groups were at clinical level in the domains of sociality, emotion and restricted interests/behavior. In the domain of communication, only one patient with Asperger's disorder had the score of 2, but all other patients in those three groups had the score of 3

or above. In the domains of communication, sociality, and emotion, group differences by post-hoc tests demonstrated a similar pattern: the three groups of autism and Asperger's had equivalent scores in these domains, while the two groups of ADHD scored equivalently. Among the three domains of communication, sociality, and emotion, scores in sociality were higher than scores in the other two domains across the groups. In this domain, the average score was around 4 in the groups of autism and Asperger's, indicating the need of considerable assistance from everyone concerned with them. Patients with PDDNOS still needed assistance to some degree at the level of 3, as considered by the leaders or supervisors of their groups. Patients with ADHD had weakness on sociality, but the level of the disturbance was in the range in which they did not need special support.

The degree of restricted interests/behaviors was low in the group with combined-type ADHD, but they still had symptoms to some extent. Half of the patients with inattentive-type ADHD were at clinical level in this domain. The degree in the group with inattentive-type ADHD was slightly higher than that of the group with combined-type ADHD, but this difference was not significant ($p = 0.482$). Also, sensory abnormality and gross and fine motor disabilities were infrequent in the group of combined-type ADHD. None of them were at clinical level in these domains. Variances

among individuals on these domains were large in all the groups. That is, although some patients suffered much from those features, the mean scores were relatively lower than those in the features such as sociality, communication, and restricted interests/behaviors. Stereotyped/repetitive motion also varied greatly between individuals, but on average was higher in the groups with autism.

Inattention, hyperactivity and impulsivity are known as symptoms of ADHD. However, the other groups also had high scores in those features. The mean scores of inattention were above 3 in all the groups. Also, in the domain of impulsivity, every group had a mean score higher than 2.5 and we did not find group differences in a post-hoc test by the Tukey method, although variation between subgroups by one-way ANOVA was detected ($p = 0.039$). In the domain of hyperactivity, the group of autism with MR had as high a score as the group of combined-type ADHD. Other groups had relatively low scores. However, more than 40 percent of other PDD patients were hyperactive at the clinical level, whereas most of the patients with inattentive-type ADHD were not hyperactive, as expected from the diagnostic definition. Variances in sleep cycle were large and group differences were not found by one-way ANOVA ($p = 0.362$).

In the domain of learning, only the group of autism without MR had a

relatively high score, 2.70 on average. This group showed a significantly higher score than the other groups except for autism with MR. Language development was delayed in the two groups of autism, as expected from the diagnostic definition. Each of these two groups had significantly higher scores than each of the remaining four groups.

4. Discussion

The MSPA was developed with the aim of understanding the entire profile of patients with PDD or ADHD and to facilitate the support that they need more directly. For diagnostic purposes, the assessment tools for these disorders already exist. Many questionnaires have been published and used for each purpose. Questionnaires are useful for gathering information from multiple observers, but there is disagreement between observers especially with psychological and behavioral symptoms as compared with physical symptoms (Marteleto, Lima e Menezes, Tamanaha, Chiari, & Perissinoto, 2008). Thus, questionnaires would be useful for screening, but not adequate for objective assessment of behavioral characteristics.

The currently available objective scales for professional assessment are listed following. The Childhood Autism Rating Scale (CARS) is widely used in clinical practice (Schopler, Reichler, DeVellis, & Daly, 1980) and recently the second edition

has been published. The Autism Diagnostic Interview-Revised (ADI-R) (Lord, Rutter, & Couteur, 1994), Autism Diagnostic Observation Schedule (ADOS) (Lord et al., 1989) and the Diagnostic Interview for Social and Communication Disorders (DISCO) (Wing, Leekam, Libby, Gould, & Larcombe, 2002) are also well known. However, each tool has its own purpose. All the previous four scales are tools for assisting with diagnosis as the names suggest. The advantages of ADI-R, ADOS and DISCO are with high validity for the purpose of diagnosis. ADI-R consists of 93 items and DISCO has 319 items. ADOS is easier to administer than the other two, but still takes about one hour. Because they are well-structured and take time to administer, they are usually used in research settings rather than in clinical practice (Miles, McCathren, Stichter, & Shinawi, 2010). CARS is as convenient as the present scale and consists of 14 items. However, this is also for the diagnosis of childhood autism, and is formulated so that all the items are correlated and the total score from 14 items indicates the degree of autism (Schopler et al, 1980). That is, the 14 items do not reflect the separate domains. CARS is restricted to children and also CARS, ADI-R and ADOS are intended to assess mainly the core features of autism or PDD. Therefore, they do not always encompass other associated features as independent domains, although each scale includes some of them. The present scale is not only for the assessment of core features of autism or PDD, but also

for ADHD, DCD and other associated features with treatment implications.

The most important difference of the present scale from the previous tools is its purpose. The purpose of this scale is not for diagnosis but for understanding behavioral characteristics and difficulties in life. From the standpoint of daily life, even if these patients and their families consult the professionals and receive the diagnosis, their difficulties would continue as long as their living situations do not correspond to their needs by meeting them adequately. When the patients are adolescents or adults, fostering insight or self-understanding would be desirable as well. The diagnosis name with ‘disorder’ might lower self-esteem (Humphrey & Lewis, 2008), but our chart can be fed back directly to patients and their families at a glance, reflecting needs to be met rather than deficits identified. Since visual cues are known to help recognition in PDD patients (Mesibov & Howley, 2003), the radar-chart format is designed to facilitate insight or self-understanding. Indeed, in our experience, this chart has led to improved insight, to building relationships of mutual trust between patients and psychiatrists, and to patients’ acceptance of their diagnosis. Furthermore, it has helped to foster shared understanding of the difficulties between the patients themselves and those in their surroundings.

In addition, the diagnosis alone would be insufficient to represent the entire

profile of dysfunction in this group of patients, partly because PDD and ADHD cannot be diagnosed simultaneously in the current diagnostic systems of the International Statistical Classification of Diseases (ICD-10) (WHO, 1992) and DSM-IV-TR, and partly because individual differences still remain even in the same diagnosis. Furthermore, the diagnostic criteria of PDDNOS itself are not fully defined (Myhr, 1998). Not only are the clinical features of these disorders multimodal, but the degree of each feature varies. Therefore, current researchers tend to regard the overall syndrome as a spectrum rather than separate subtypes (Szatmari, 1992). However, the spectrum concept is insufficient to capture the individual features of these patients, and the assessment of the level of each behavioral feature is very important for daily life and social and environmental adaptation. Therefore, we designed this multimodal and nine-stepped representation which is also useful for mild or high-functioning cases. Taken together, we believe that the present scale adds value for treatment beyond the above excellent tools aimed chiefly at diagnosis.

The reliability of the present scale was adequate among trained psychiatrists. However, some training would be needed in raters. Therefore, we are in the process of estimating the amount of training required to increase the number of specialists who can reliably use this assessment. We used the present scale mostly for the outpatients in the

department of psychiatry. Therefore, we assessed not only young children but also adolescents and adults. It was very useful for adolescents and adults for understanding themselves. For children, caregivers mainly referred to this scale to support them. The assessment using this scale is based on long-term characteristics, that is, we gathered information from birth throughout development, not limited to the present state. Therefore, the scores are stable and we assume that age does not affect them much when the information is adequate. On the contrary, this scale is not suitable to assess short-term changes such as the effects of intervention. We do not assume that the present scale captures adaptation by intervention or temporary symptoms in exacerbation. However, we are planning a longitudinal study to capture long-term changes of characteristics beyond superficial appearances.

When we consider each domain, sociality is the main domain that divides groups as expected. The outcome that patients with ADHD still had some social difficulty below the clinical level is consistent with a previous report (Nijmeijer et al., 2008). We also showed that inattention was the one domain or feature where all the groups had difficulties at clinical level. Similarly, Strum et al (2004) reported that 95 out of 101 children with PDD had attention deficit. Thus, we might consider the existence of inattention even in groups other than ADHD, because inattention hinders adaptation

in various aspects of daily life. Inattention might be a common feature not only in ADHD but also in PDD. The domain where further consideration is required before a conclusion can be made would be the sleep cycle. Richdale and Schreck (2009) reported that sleep problems are often found in PDD and ADHD, but are multifactorial. They may be an innate feature or may be from stress due to difficulties in life. We will try to detect the innate part and aim to modify the present scale.

Regarding group differences, our results showed that the main difference between the autism and Asperger groups was language development, as defined by the diagnostic criteria. Symptom severity in the Asperger group was milder in the domains of sensory, stereotyped/repetitive motion, and motor skills, but without significant differences (see Fig. 1b). As to the PDDNOS group, symptom severity was even milder than the Asperger group in domains listed as the diagnostic criteria in DSM-IV-TR (communication, sociality, emotion, restricted interests/behaviors, and stereotyped/repetitive motion), but did not differ in other domains such as motor skills, ADHD symptoms, learning and language development. That is, the difference of features among these three groups is mainly in the domains which constitute the diagnostic definitions of them. Therefore, these three subtypes of PDD are supposed to be a continuum, although they are divided by the diagnostic criteria. Interestingly,

features in the group of inattentive-type ADHD were even milder but as a pattern of symptoms, they were similar to those in the group of PDDNOS. This suggests that PDD and ADHD are also related as many previous reports have pointed out (Jensen et al, 1997; Yoshida & Uchiyama, 2004; Frazier & Youngstrom, 2006; Lee & Ousley, 2006; Nijmeijer et al, 2009; Sinzig et al, 2009; Kochhar et al., 2010), indicating the need for a shared assessment tool for PDD and ADHD.

Of interest, only the group of combined-type ADHD showed a different pattern. This group had low scores in the domains of restricted interests/behavior, sensory, and motor skills. Pitcher et al (2003) also reported that hyperactive/impulsive type had better motor skills than inattentive type. These results identify the possibility that hyperactivity might have positive consequences, such as improved motor skills. These group differences and domain analyses might give novel insights on biological mechanisms of these disorders after further consideration.

5. Conclusions

We developed a multi-dimensional and quantitative assessment chart for PDD and ADHD. The reliability among trained psychiatrists was sufficient. This tool uncovers multi-dimensional clinical features of each patient at a glance, and is useful for

fostering understanding of these difficulties by patients themselves and also by those in their surroundings. Furthermore, it discloses the relationships and the differences among subtypes of PDD and ADHD. We hope that it will help patients with these disorders and the clinicians who treat them and also become a useful tool in research settings investigating the biological backgrounds of these disorders.

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Tables

Table 1. Group characteristics

	Autism with MR	Autism without MR	Asperger	PDDNOS	ADHD combined	ADHD inattentive
Number	21	18	40	74	12	14
Male:Female	16:5	15:3	29:11	59:15	9:3	9:5
Age	12.3 ± 8.0	14.1 ± 12.5	14.2 ± 8.6	14.7 ± 11.5	13.3 ± 6.9	14.7 ± 8.5
FIQ	51.6 ± 14.3 ^a	87.3 ± 13.0	91.8 ± 20.3	94.8 ± 17.7	96.3 ± 8.3	95.5 ± 17.6
VIQ	47.1 ± 18.8 ^a	90.2 ± 16.6	93.8 ± 21.9	96.2 ± 19.3	96.8 ± 11.0	98.5 ± 22.3
PIQ	53.2 ± 16.7 ^a	85.7 ± 14.6	91.0 ± 20.2	94.3 ± 16.9	96.2 ± 9.4	91.6 ± 13.4

^a Significantly different from other groups

Table 2. Results of the inter-rater reliability

Domain	ICC
Communication	0.845
Sociality	0.877
Emotion	0.849
Restricted interests/behaviors	0.964
Sensory	0.959
Stereotyped/repetitive motion	0.961
Gross motor	0.975
Fine motor	0.834
Inattention	0.947
Hyperactivity	0.983
Impulsivity	0.973
Sleep cycle	0.949
Learning	0.975
Language development	0.966

Table 3. Percentages of patients at clinical level in each group by domains

	Autism with MR	Autism without MR	Asperger	PDDNOS	ADHD combined	ADHD inattentive
Communication	100	100	97.5	54.1	0	21.4
Sociality	100	100	100	82.5	16.7	28.6
Emotion	100	100	100	58.2	16.7	14.3
Restricted interests/behaviors	100	100	100	83.8	16.7	50
Sensory	57.2	66.7	40	27.1	0	14.2
Stereotyped/ repetitive motion	42.9	39	25	9.5	8.3	0
Gross motor	47.6	47.1	23.1	23.3	0	14.2
Fine motor	47.7	29.4	12.8	12.5	0	21.4
Inattention	71.3	77.8	75	64.2	100	100
Hyperactivity	71.3	44.6	40	46	100	0
Impulsivity	71.4	53	62.5	51.4	91.7	42.8
Sleep cycle	28.6	16.7	17.9	18.1	18.2	28.5
Learning	22.2	60	16.2	14.7	9.1	14.3
Language development	100	100	0	13.2	0	7.7

We regarded the clinical level as the score of 3 or above. We calculated each percentage of such patients in each group by domains.

Figure Captions

Figure 1. Radar-chart representation

a. Case examples: We showed three cases as examples on a radar-chart. Scores outside red broken lines mean that special supports are required. Case 1 and 2 had the same diagnosis, Asperger's disorder, but the clinical features were quite different. Case 1 had both symptoms of PDD and ADHD. Case 2 suffered from repetitive motion and clumsiness. Case 3 was diagnosed as PDDNOS and had sensory abnormality and learning disabilities.

b. Mean scores by groups: We showed the mean scores of six diagnostic groups on a radar-chart. Color lines and symbols are explained in the inset. All the groups suffered from inattention. Inattentive-type ADHD had similar but milder features compared with PDDNOS. Combined-type ADHD had a different pattern from other groups. That group had low scores in the domains of restricted interests/behaviors, sensory and motor skills.

Figure 2. Group differences in each domain

Bars indicate the mean scores and the error bars are the standard deviations. ★ and ★ mean that the differences were significant at $0.01 < p < 0.05$, $p \leq 0.01$,

respectively. In the domains of communication and sociality, we only showed NS where the group differences were not significant, because most pairs showed significant differences. In other domains, all the significant differences are shown by the above signs.

Communication



